

Unusual Pain due to Unilateral Facet Degeneration at the C1-2 Level

C1-2 Unilateral Faset Dejenerasyonuna Bağlı Nadir Görülen Ağrı: Olgu Sunumu

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ABSTRACT

AIM: Neck pain is a common problem that may result from abnormalities in the soft tissues, bones and joints of the spine. However, upper neck and occipital pain syndrome due to unilateral idiopathic arthrosis of a C1-2 lateral articulation is a very rare condition.

MATERIAL and METHODS: We describe two patients with unilateral arthrosis of a C1-2 lateral articulation who were successfully treated with C1-2 fusion.

RESULTS: The patients presented with severe headache and/or neck pain. The diagnosis was made with a careful history and imaging studies, including X-ray films, CT scans, and MRI studies. Posterior atlanto-axial transfacet screw fixation was performed in both patients. There were no surgical complications.

CONCLUSION: C1-2 fusion is necessary to relieve intractable pain after failure of conservative treatment and/or invasive pain procedures in unilateral C1-2 arthrosis.

KEYWORDS: Unilateral arthrosis, Cervical, Pain syndrome, Fusion

ÖΖ

AMAÇ: Boyun ağrısı omurgada eklem, kemik ve yumuşak dokudaki patolojilere bağlı olarak çok sık görülen bir bulgudur. Ancak C1-2'deki faset eklemlerinin tek taraflı harabiyetine bağlı ağrı oluşumu son derece nadirdir.

YÖNTEM ve GEREÇLER: C1-2 unilateral faset dejenerasyonuna bağlı ağrılı iki olgu sunulmuştur.

BULGULAR: Hastalar konservatif tedaviye yanıtsız olduğu için cerrahi tedavi yapılmıştır. Unilateral harabiyet X-ray, CT ve MR ile tespit edilmiştir. Cerrahi olarak posterior atlantoaksiyal transfaset vida fiksasyonu uygulanmıştır. Cerrahi komplikasyon gelişmemiştir.

SONUÇ: C1-C2 ünilateral faset dejenerasyonunda konservatif tedavi ve/veya invazif ağrı prosedürlerinin başarısız olduğu durumlarda C1-C2 füzyon ameliyatı ağrıyı geçirmek için gereklidir.

ANAHTAR SÖZCÜKLER: Unilateral artrozis, Servikal, Ağrı sendromu, Füzyon

INTRODUCTION

It is estimated that the yearly incidence of neck pain ranges from 10.4% to 21.3% (13).

There are a number of differential diagnoses of this type of pain, including cervical intervertebral disc degeneration, trauma, fibromyalgia, tumors and psychogenic pain disorders (9,19).

In two separate studies, the prevalence of facet joint involvement was estimated as 33-42% (19) and 35-40% (20). Multiple components can be the source of pain, including the fibrous capsule, the synovial membrane or the hyaline

cartilage surface. There are also free nerve endings and nociceptors, (21) and the release of the pain mediators substance P, neuropeptide Y and the calcitonin gene-related peptide (15).

Degeneration of the articulating surfaces marks an aging process and is therefore believed to have a high prevalence in the population; however, the exact prevalence of this disorder remains unknown (16).

In this report, we present very rare cases of unilateral C1/C2 facet joint degeneration in young patients, which have not been well described in the literature.

CASE REPORTS

Patient A: A 51-year-old woman presented with severe headaches that had been increasing daily over a two-year span. Physical examination revealed aggravation of the pain while rotating the head, whereas the main source of pain was located at the base of the cranium. There were no pathologic neurological signs to be found. Magnetic resonance(MR), CT and X-Ray studies showed massive degeneration of the right superior articular surface in C2 (Figure 1A, B). In addition, there was no history of a rheumatic disorder. Minimal invasive pain procedures and drug treatment did not relieved the pain.

Due to the persistence of the symptoms for a six-month period, a posterior atlanto-axial transfacet screw fixation (Magerl Technique) surgery was performed (Figure 1C,D). After treatment, the patient had a good outcome, as confirmed at the 2-year follow-up examination.

Patient B: This patient was a 54-year-old woman with a 6-month history of upper neck pain. Her symptoms were severely affecting her head movement. The origin of the pain was at the base of the cranium and the pain increased while rotating the head. Neurologic examination was normal. CT, X-Ray and MR scans showed degeneration of the left inferior articular surface of the C1 (Figure 2A,B). She did not benefit from medicine for 6 months and did not accept percutaneous injections to the C1-C2 region.

She underwent posterior atlanto-axial transfacet screw fixation (Magerl Technique) (Figure 2C,D). This treatment

resulted in a favorable patient outcome, as seen at the followup after two years.

DISCUSSION

Lifetime prevalence of neck pain is reported to be between 26% and 71%. It has been well accepted that it has a significant impact on the economy, society, and the health of the working population (4,12,13).

Cervical facet or zygapophysial joints can be a source of pain in the neck and referred pain in the head and upper extremities (2,7,22). It has been shown that the cervical facet joints are well innervated by the medial branches of the dorsal rami with free and encapsulated nerve endings with nociceptors and mechanoreceptors (3,5,15,21). There are several different grading systems for cervical facet joint degeneration, (17,24) but unfortunately none has become the gold standard. Hence, to determine the necessary treatment, it may be more important to evaluate the degree of patient complaint than the degree of degeneration observed during the radiological investigation. Clinical symptoms often include palpatory pains over the affected joint or the adjacent muscles (11) and/or pain when rotating or extending/flexing the head. In our case series, the major complaint of the patients was a persistent headache and neck pain, especially with movement of the head. The pain was not relieved despite long-term medication and an invasive pain-relief procedure. We agree with the hypothesis that the cervical zygapophyseal joints and the compression of medial branches of the dorsal



Figure 1: A) Coronal and **B)** axial computed tomography scans of the upper cervical spine showing arthrotic enlargement and joint space narrowing on the right (arrows). **C)** Lateral and **D)** antero-posterior radiograph of the cervical spine following surgery showing the C1-2 instrumentation.



Figure 2: A) Coronal and B) axial computed tomography scans of the upper cervical spine showing arthrotic enlargement and joint space narrowing on the left (arrows). C) Lateral and D) antero-posterior radiograph of the cervical spine following surgery showing the C1-2 instrumentation.

rami are the source of pain in these patients (23). Cervical spondylosis usually produces intermittent neck pain with stiffness. The pain is usually chronic and episodic and may spread to the occiput and shoulder (20). In addition, nearly one-third of the patients experience headache. Pain may also spread to the shoulders, upper arm, forearm or hand in approximately two-thirds of the patients (10). However, patients with cervical spondylosis that involves the upper cervical spine often experience neck and shoulder pain and occasionally suboccipital headache. The possible sources of cervicogenic headache are a degenerated atlanto-occipital joint and atlanto-axial joints. C1 - C3 dermatomes participate in the innervation of the back of the head and the neck muscles. Moreover, these joints are not innervated by the cervical dorsal rami but receive branches from the C1 - C2 ventral rami. It has been suggested that the pain suffered by patients arises not from compression of the nerve, but as a primary irritation in the joint itself (8). However, it should be kept in mind that pain due to cervical spondylosis may radiate/spread to the suboccipital region, skull base and vertex (23).

The atlanto-axial joint is responsible for up to 75% of the head's rotation. When taking the patient's anatomy into consideration, one will notice the comparatively large articulating surfaces of the diarthrodial lateral mass articulations. Along with the medial atlanto-axial joint, these represent a functional unit that provides mechanical stability and ensures physiological flexion, extension and rotation of the head (14). Morphometric studies have shown good congruency in the sagittal plane between the two articulating surfaces under physiological conditions, (5) with the largest rotation in the horizontal plane, (25) During rotation, the C1 and the C2 do not move at the same speed. In addition to the fact that synovial fluid is also diminished and loses, contacting surface could be a possible reason why pain increases during rotation. Degeneration of the articulating surface occurs as a result of multiple factors, such as injury, aging, infections and genetic disposition.

Degeneration of the upper cervical spine may be observed in various rheumatologic or neurological disorders and experienced bilaterally (1,10,18). However, unilateral idiopathic degeneration of the joints such as in our cases is uncommon. Generally, degeneration of the surfaces of the joint occurs quite evenly (16).

C1-2 fusion is necessary to relieve intractable pain after the failure of conservative treatment and/or invasive pain procedures. Hence we performed C1-2 fusion in our patients, and this treatment provided permanent relief to the two patients.

An alternative surgical technique is C1 lateral mass and C2 pedicular instrumentation. This technique offers satisfactory stabilization.

In summary, unilateral idiopathic arthrosis of a C1-2 lateral articulation is a very rare condition. Cervical spondylosis should be considered for the differential diagnosis. Patients who continue to experience severe pain despite other treatments may require detailed diagnostic studies. Cases with intractable pain require surgery for the elimination of motion at the pathological sites.

REFERENCES

- 1. Angelini L, Broggi G, Nardocci N, Savoiardo M: Subacute cervical myelopathy in a child with cerebral palsy. Secondary to torsion dystonia? Childs Brain 9:354–357, 1982
- Aprill C, Dwyer A, Bogduk N: The prevalence of cervical zygapophyseal joint pain patterns II: A clinical evaluation. Spine 15:458-461, 1990
- 3. Bogduk N: The clinical anatomy of the cervical dorsal rami. Spine 7:319- 330, 1982
- Bot SD, van der Waal JM, Terwee CB, van der Windt DA, Bouter LM, Dekker J: Incidence and prevalence of complaints of the neck and upper extremity in general practice. Ann Rheum Dis 64:118-123, 2005
- Cattrysse E, Provyn S, Gagey O, Kool P, Clarys JP, Van Roy P: In vitro three dimensional morphometry of the lateral atlantoaxial articular surfaces. Spine 33:1503-1508, 2008
- Chen C, Lu Y, Kallakuri S, Patwardhan A, Cavanaugh JM: Distribution of A-delta and C-fiber receptors in the cervical facet joint capsule and their response to stretch. J Bone Joint Surg Am 88:1087-1816, 2006
- Dwyer A, Aprill C, Bogduk N: Cervical zygapophyseal joint pain patterns: A study in normal volunteers. Spine 15: 453-457, 1990
- 8. Ehni G, Benner B: Occipital neuralgia and the C1-2 arthrosis syndrome. J Neurosurg 61(5):961-965, 1984
- Fletcher G, Haughton VM, Ho KC, Yu SW: Age-related changes in the cervical facet joints: Studies with cryomicrotomy, MR, and CT. AJR Am J Roentgenol 154:817-820, 1990
- Fuji T, Yonenobu K, Fujiwara K, Yamashita K, Ebara S, Ono K, Okada K: Cervical radiculopathy or myelopathy secondary to athetoid cerebral palsy. J Bone Joint Surg 69:815–821, 1987
- Fukui S, Ohseto K, Shiotani M, Ohno K, Karasawa H, Naganuma Y, Yuda Y: Referred pain distribution of the cervical zygapophyseal joints and cervical dorsal rami. Pain 68:79-83, 1996

- 12. Hogg-Johnson S, van der Velde G, Carroll LJ: Bone and joint decade 2000-2010 task force on neck pain and its associated disorders. The burden and determinants of neck pain in the general population: Results of the bone and joint decade 2000- 2010 task force on neck pain and its associated disorders. Spine 33:39-51, 2008
- 13. Hoy DG, Protani M, De R, Buchbinder R: The epidemiology of neck pain. Best Pract Res Clin Rheumatol 24:783-792, 2010
- 14. Jaumard NV, Welch WC, Winkelstein BA: Spinal facet joint biomechanics and mechanotransduction in normal, injury and degenerative conditions. J Biomech Eng 133(7):071010, 2011
- 15. Kallakuri S, Singh A, Chen C, Cavanaugh JM: Demonstration of substance P, calcitonin gene-related peptide, and protein gene product 9.5 containing nerve fibers in human cervical facet joint capsules. Spine 29:1182-1186, 2004
- Kettler A, Werner K, Wilke HJ: Morphological changes of cervical facet joints in elderly individuals. Eur Spine J16: 987-992, 2007
- Kettler A, Wilke HJ: Review of existing grading systems for cervical or lumbar disc and facet joint degeneration. Eur Spine J 15:705-718, 2006
- Loher TJ, Barlocher CB, Krauss JK: Dystonic movement disorders and spinal degenerative disease. Stereotact Funct Neurosurg 84:1–11, 2006
- Manchikanti L, Manchikanti KN, Cash KA, Singh V, Giordano J: Age-related prevalence of facet-joint involvement in chronic neck and low back pain. Pain Physician 11:67-75, 2008
- 20. Manchikanti L, Manchikanti KN, Pampati V, Brandon DE, Giordano J: The prevalence of facet-joint-related chronic neck pain in postsurgical and nonpostsurgical patients: A comparative evaluation. Pain Pract 8:5-10, 2008
- 21. McLain RF: Mechanoreceptor endings in human cervical facet joints. Spine 19:495-501, 1994
- 22. Pawl RP: Headache, cervical spondylosis, and anterior cervical fusion. Surg Ann 9:391-498, 1977
- 23. Resnick D: Hyperostosis and ossification in the cervical spine. Arthritis Rheum 27:564–569, 1984
- 24. Walraevens J, Liu B, Meersschaert J, Demaerel P, Delye H, Depreitere B, Vander Sloten J, Goffin J: Qualitative and quantitative assessment of degeneration of cervical intervertebral discs and facet joints. Eur Spine J 18:358-369, 2009
- 25. Zhang H, Bai J: Development and validation of a finite element model of the occipito-atlantoaxial complex under physiologic loads. Spine 32:968-974, 2007